A rare case of oral melanoacanthoma

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Abstract

Oral melanoacanthoma is a rare, pigmented lesion which brings suspicion of a malignancy with its sudden appearance and rapid growth. It is characterized clinically as a brown-black lesion and histologically it shows acanthosis of the superficial epithelium and proliferation of dendritic melanocytes. Due to their unusual clinical pattern, biopsy is mandatory to rule out malignancy.

Keywords: Melanoacanthoma, melanoma, oral pigmentation

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INTRODUCTION

Melanoacanthoma is an uncommon, benign, mucocutaneous pigmented lesion characterized by dendritic melanocytes dispersed throughout the acanthotic epithelium.^[1] Oral melanoacanthoma is a rare condition indicative of a reactive process.^[2] The first case of oral melanoacanthoma was reported in 1978 by Tomich.^[1]

CASE REPORT

A 55-year-old female patient reported to the Department of Oral Medicine and Radiology with a chief complaint of pigmentation on the buccal mucosa of both sides and labial mucosa of both upper and lower lips for 4 months. The patient was apparently asymptomatic 4 months back. There was no associated symptom or any discomfort. Pigmentation rapidly increased to the present size [Figure 1]. The patient did not have any habit history. Extraoral examination revealed no relevant findings. Intraoral examination revealed diffuse, multiple macular black pigmentation on the upper and lower labial mucosa and

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bilaterally on the buccal mucosa. Lesions were irregular in shape with no ulceration or growth. On palpation, no raised surface, nodularity, tenderness, fixation to the underlying mucosa, and blanching of the lesions could be found.

An incisional biopsy from the left buccal mucosa was performed and sent for histopathological examination. Gross specimen comprised a soft tissue measuring 5 mm × 4 mm × 4 mm in size, irregular in shape, brownish in color, and soft in consistency. A routine hematoxylin and eosin (H&E) staining was performed with an additional subsequent staining of one slide with Fontana-Masson silver impregnation stain.

The H&E-stained section showed nonkeratinized stratified squamous epithelium. There was marked acanthosis with diffuse distribution of dendritic melanocytes in suprabasal and spinous layers of the epithelium [Figure 2]. There was no cytologic atypia. A chronic inflammatory cell infiltrate was present in the subjacent connective tissue. The dendritic melanocytes were also demonstrated by Fontana-Masson stain [Figure 3]. Based on the history, clinical features,

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and histological presentation, the lesion was diagnosed with melanoacanthoma. The patient has been kept on follow-up for 6 months, and during this time, there was no change appearance and neither increase nor decrease in the size of the pigmentation.

DISCUSSION

After the first case of Tomich, a total of 65 cases have been documented. [1] The prevalence of oral melanoacanthoma in the Indian population is not known. Our literature search revealed a total of six cases.

Oral melanoacanthoma is a reactive, benign, uncommon acquired pigmentation of the oral mucosa characterized by



Figure 1: Intraoral photograph showing pigmentation on both labial and buccal mucosa

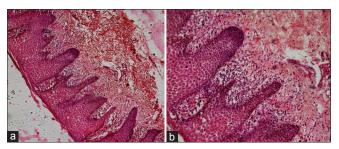


Figure 2: Histopathological image showing melanin pigmentation (H&E stain, [a] ×4, [b] ×10)

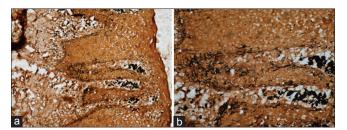


Figure 3: Histopathological image showing melanocytes and melanin pigmentation (Fontana-Masson stain, [a] $\times 20$, [b] $\times 40$)

dendritic melanocytes dispersed throughout the epithelium. It is unrelated to the melanoacanthoma of the skin. [3] It is predominantly observed in blacks though occurrence has also been reported in Caucasians, Hispanics and Asians.^[2] There is a female predilection with a male-to-female ratio of 2:1. The most common age of incidence is reported to be the third and fourth decades of life with an age range of 9-77 years.^[1-3] Buccal mucosa is the most common site of involvement, while other sites such as the mucosa of the lip, palate, gingiva and alveolar mucosa have also been reported.^[2,3] Clinically, the lesion appears to be asymptomatic, flat or slightly raised black or brown macule of few millimeters to several centimeters in size. [2,3] The lesions are usually solitary and well circumscribed though bilateral and multiple presentations are also reported. The lesion may grow rapidly thus bringing suspicion of malignancy. [2] In our case, the lesion had a diffuse presentation and increased rapidly in size.

The lesion is clinically indistinguishable from many other oral pigmented lesions. All pigmented lesions should be followed up and observed for any change in size, shape, color, surface, or any other symptom over time. Any such feature mandates a biopsy to rule out a malignancy. Oral melanoacanthoma rapidly increases in size and can reach a diameter of several centimeters within a few weeks. This rapid growth of oral melanoacanthoma may mimick malignant melanoma, especially the radial growth phase of an *in situ* melanoma. The biopsy is the only way to rule out the possibility a melanoma.

Although the etiology is not clearly known, it is largely attributed to local irritation or even mild trauma.^[2] The clinical differential diagnosis for oral melanoacanthoma may include other pigmented mucosal lesions such as physiologic pigmentation, pigmentation due to smoking and medication, oral nevi, focal melanosis, and most importantly, melanoma. However, an accurate history can exclude these entities, as only oral melanoacanthoma exhibits a rapid growth in size during a period of only a few weeks. However, in the absence of such a history, the histologic examination provides the definitive diagnosis.^[4] The diffuse oral pigmentation can also be seen in other conditions such as Peutz–Jeghers syndrome, Addison's disease and Laugier–Hunziker syndrome.^[5]

Diffuse pigmentation of the oral mucosa and/or skin secondary to systemic drug administration is a well-recognized phenomenon. [5] The patient did not give any history of medications including oral contraceptive, steroids and antimalarial drugs.

Peutz–Jeghers syndrome is an autosomal dominant disorder with variable inheritance, characterized by hamartomatous polyps in the gastrointestinal tract, mostly in the small intestine and pigmented mucocutaneous lesions.^[5] In the present case, the patient had no previous or family history of any kinds of pigmented lesions or gastrointestinal polyposis.

In Addison's disease, reactive mucosal hyperpigmentation may be a common finding, since deficiency of cortisol leads to increased production of adrenocorticotropic hormone, a pituitary hormone with a chemical structure similar to that of melanocyte-stimulating hormone, stimulating the pigmentation of the skin and mucosa. Hyperthyroidism may also be associated with hyperpigmentation. These hormonal effects may enhance the proliferation and activation of melanocytes causing melanocytic hyperplasia and deposition of melanin. [6] In the present case, the patient's hematological examination reports did not reveal any hormonal abnormality.

Laugier-Hunziker syndrome is an idiopathic condition, commonly seen in white population, showing macular brownish-black pigmentation of the skin and oral mucosa including lips associated with longitudinal melanonychia of nails.

Microscopically, numerous dendritic melanocytes are seen scattered throughout the epithelium.^[2,5] Basal layer melanocytes may be increased in number, and spongiosis is a common finding. [4] The adjacent connective tissue stroma shows chronic inflammatory cell infiltrate. There may be the presence of eosinophils but that is not an essential feature for the diagnosis of oral melanoacanthoma.^[2] Diagnosis of oral melanoacanthoma can be made solely on the basis of histological features and special staining. Fontana-Masson silver impregnation technique can be used to demonstrate dendritic melanocytes and emphasize the presence of melanin.^[1] Immunohistochemical analysis shows the positive immunoreactivity of the melanocytes for S100, Melan-A/Mart-1, HMB45 and tyrosinase. [2] However, strong reactivity to HMB45 and S100 is also seen in malignant melanoma.[1]

It is a benign condition and no treatment is required once the diagnosis is confirmed. Some cases regress spontaneously or following incomplete removal or elimination of local irritant. In our case, the patient has been kept on regular follow-up for 6 months and the lesion remained unchanged during that time.

CONCLUSION

Oral melanoacanthoma is a rare condition which must be distinguished from other pigmentation. Its rapid progression warrants biopsy to rule out malignancy. Treatment is usually not required except removal of all local causes of trauma.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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